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Case report

Nodular fasciitis in the masticator space eroding into the mandible: a case report

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ABSTRACT

Nodular fasciitis is a benign soft tissue neoplasm of mesenchymal origin. It is usually characterized by rapid growth, infiltrative behavior, and heterogeneous histopathology, which can make diagnosis difficult and lead to delayed management. It has a 15%-20% occurrence rate in the head and neck and occurs rarely intraorally. In this report, we discuss an unusual case of nodular fasciitis originating in the masticator space and destroying the ascending ramus of the mandible. The treatment involved complete resection of the lesion and reconstruction with a temporomandibular joint prosthesis. At 24 months after surgery, the patient showed a return to normal function with no signs of recurrence.

1. Introduction

Nodular fasciitis (NF) is a soft tissue proliferation of mesenchymal origin consisting of fibroblasts and myofibroblasts [1]. It often presents clinically with aggressive features including rapid growth, destruction of adjacent anatomic structures, and heterogeneous histopathology, making diagnosis difficult and delaying appropriate and timely treatment. It most commonly occurs in the upper extremities (48%), the trunk (20%), head and neck (15%-20%), and followed by the lower extremities (15%). It can occur in all ages, but most commonly in adults between ages 20-40 years [2]. Inflammatory and traumatic causes have been implicated, with trauma as a trigger in 10%-15% of cases [3]. Moreover, recent molecular cytogenetic studies identified the presence of USP6 fusion genes in NF lesions confirming a clonal neoplastic origin [4]. USP6 is a ubiquitin-specific protease involved in numerous cell processes, expressed primarily in testicular tissue, and has been shown to have oncogenic activity [4]. This favors NF as a neoplastic growth and not as a reactive inflammatory lesion.

In this case report, we will present and discuss the management of an unusual case of NF of the masticator space eroding the ascending ramus of the mandible.

2. Presentation of case

A 37-year-old man presented to the oral and maxillofacial surgery clinic at the Montreal General Hospital with left preauricular swelling of 1-month duration. The patient is otherwise healthy, does not take any medication, has no allergies, is nonsmoker, and has no family history of malignant disease. On examination, the patient was afebrile, had no constitutional symptoms, and experienced no recent significant weight loss. The neck range of movement was within normal limits, there was no neck swelling or lymphadenopathy, and the facial nerve function was normal bilaterally. The mouth opening was limited to 20 mm and has been worsening over a 1-month period. There was no clicking or crepitus on temporomandibular joint (TMJ) palpation. A left preauricular firm and fixed nonmobile mass of approximately 2 × 2 cm was present with no associated overlying skin changes (Figure 1). Intraorally, the airway was intact, and a firm submucosal swelling was palpable in the left

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Figure 1. Preoperative frontal photograph showing left preauricular mass.

masticator space with no associated erythema or signs of infection. Computed tomography and magnetic resonance (MR) images were taken, which confirmed the presence of a well-defined homogeneous mass measuring 5.2 × 4.5 × 3.2 cm in the left masticator region associated with destruction of the left ascending ramus and condyle (Figure 2). An ultrasound-guided biopsy was performed, but the analysis of the specimen was inconclusive showing mildly cellular spindle cell proliferation of myofibroblastic nature.

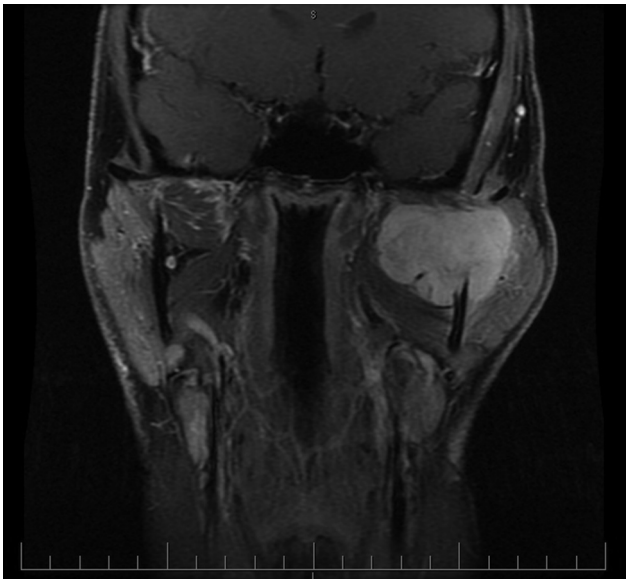

Figure 2. Preoperative imaging. Magnetic resonance imaging coronal view (T1-weighted image post-gadolinium contrast) showing the mass with uptake of contrast agent in the left masticator region.

The limitation in mouth opening continued to worsen rapidly over few days, and the lesion became associated with an intraoral firm and ulcerated swelling with rolled borders in the left posterior buccal mucosa. An intraoral incisional biopsy was obtained under general anesthesia, and the diagnosis was confirmed as NF. Intra-op, an initial injection of triamcinolone (10 mg) was performed into the center of the lesion after frozen section confirmation and was then followed with another injection of the same amount 6 weeks later using ultrasound guidance, with no significant change in the size of the lesion or clinical improvement. At that point, it was decided to resect the lesion and the involved structures and plan for reconstruction. A vertical ramus compartment resection was performed including resection of the mandibular condyle [5]. A custom-made TMJ prosthesis was used to reconstruct the defect. The postoperative care was uneventful, and 24 months later, the patient has good range of mandibular motion and esthetics with no evidence of recurrence on MR imaging (Figures 3 and 4). The post-operative panoramic x-ray shows stable hardware at 2 years (Figure 5).

3. Discussion

NF lesions can originate from the subcutaneous, intramuscular, or fascial regions; it can also be located intravascularly or intradermally, but these latter are rather rare [2,6]. The subcutaneous type is the most common (3-10 times more common than the other types) and usually presents as a subcutaneous nodule. The

Figure 3. Postoperative photograph 24 months after surgery.

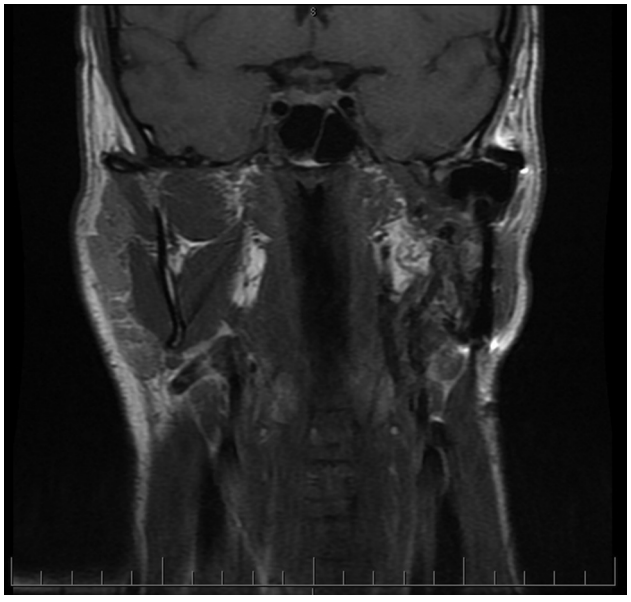

Figure 4. Postoperative imaging. Magnetic resonance imaging coronal view (T1-weighted image post-gadolinium contrast) showing left ascending ramus and temporomandibular joint prosthesis, with no evidence of recurrence.

intramuscular subtype tends to have larger size, is associated with more aggressive and infiltrative behavior, and hence, most commonly resembles a soft tissue malignancy [2,6]. The usual size at presentation for a NF lesion is <4 cm [6]. In our case report, the size was 5.2 cm in the greatest dimension.

Seven percent to 20% of NF cases occur most commonly along subcutaneous tissues adjacent to bony prominences such as the zygoma, angle of the mandible, and anterior and inferior border of the mandible [7]. Two cases were identified involving the larynx [8], 2 cases reported involving the pharynx and hypopharynx [9,10], and only 1 case reported involving the TMJ [11].

Our case is unique as it is the first case reported in the masticator space. The masticator space is bounded by fascia from the superficial layer of the deep cervical fascia and contains the medial pterygoid muscle, the lateral pterygoid muscle, a portion of the temporalis muscle, the masseter muscle, and the posterior body and ramus of the mandible. It is bounded medially by the parapharyngeal space, posteriorly by the parotid space and anteriorly by the buccal space [12]. Infections represent a more common cause of mass in the masticator space than tumors, and these are often secondary to an odontogenic source [13]. Tumors within the masticator space can arise from muscle (eg, masticator hypertrophy, hemangioma, rhabdomyosarcoma), the mandible (odontogenic cysts or tumors, osteosarcoma), nerve (eg, schwannoma), or from local extension from salivary gland tissue, the nasopharynx, and the oropharynx [13]. Also, distant metastases such as breast adenocarcinoma can be considered [13].

This case was also unique in that it showed both a deep-seated destructive lesion and intraoral manifestation in the buccal mucosa. NF in the oral region is rare—the buccal mucosa being the most common site [3]. Oral NF usually presents with an exophytic nodule with ulceration and raised margins or can be deep seated. These tend to occur between the fourth and fifth decades and show a variable period of growth ranging from several days to 2 years [3]. In this case, 11 days after ultrasound-guided biopsy, the patient presented with pain and severe trismus and intraoral ulcerated and firm swelling in the posterior buccal mucosa with rolled borders. Perhaps, the deep-reaching ultrasound-guided biopsy stimulated proliferation of the lesion.

Bone erosion in association with NF is rare. It has been reported mainly in the cranial fasciitis variant. These lesions usually present as firm and rapidly growing scalp mass in children <6 years old, have identical histopathology as NF, and can be associated with lytic bone lesions (cortical erosion) radiographically [14]. Bone erosion has also been shown in the intramuscular subtype [6] and in cases involving the appendicular skeleton [15,16]. The lesion presented in this case report most likely represents a fascial origin with ingrowth into muscle. To our knowledge, this is the first case of NF associated with bone erosion of the mandible.

Histologically, NF lesions appear as immature fibroblasts, so-called myofibroblasts, and form short fascicles and bundles, imparting a tissue culture growth pattern [1,3] (Figure 6). Other features include the presence of spindle cells admixed with loose stroma, inflammatory cells, and erythrocytes [1,17], and also mitotic activity without atypical mitosis [6]. Microscopic findings can also provide temporal information, with early lesions showing myxoid appearance progressing into cellular and then into more fibrous appearance in mature lesions [6].

Intralesional steroid use has been documented in a 34-year-old woman with a subcutaneous type NF in the volar aspect of the right forearm and showed rapid resolution [17]. In the current case, intralesional steroid injections were used when the lesion reached a peak size of 6 × 5.6 × 5.4 cm and had actually decreased to 5.2 × 3.1 × 3.5 cm in 5 weeks. However, this minimal decrease was

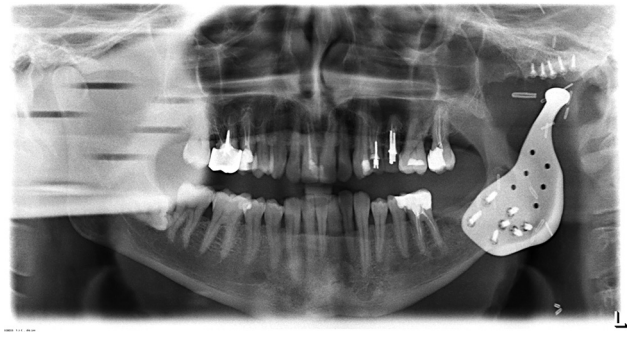

Figure 5. Post-op panorex at 2 years.

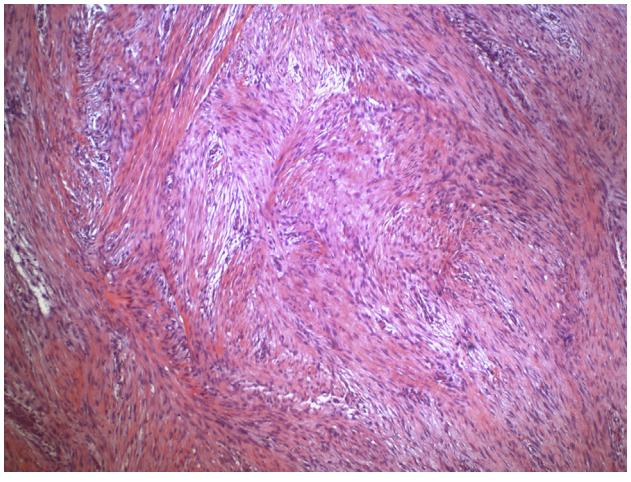

Figure 6. Histopathologic view. Microscopic image showing cellular proliferation of reactive myofibroblasts forming short fascicles and bundles imparting a tissue culture growth pattern (hematoxylin and eosin, magnification ×40).

not enough for alleviating the clinical symptoms. More research is needed regarding intralesional steroid injection and its benefit for this type of neoplasm, especially to find out whether multiple intralesional steroid injections at regular intervals would be of benefit.

4. Conclusion

The differential diagnosis for this tumor is as follows: fibrosarcoma, myofibroma, neurofibroma, solitary fibrous tumor, fibromatosis, and also fibrous histiocytoma. This diverse differential and the inclusion of aggressive malignant tumors make it difficult to distinguish these lesions on computed tomography and MR imaging [6], which makes imaging for NF helpful mainly in outlining the extent of the lesion and for surgical planning. Indeed, histopathologic diagnosis is still the best method for diagnosis and has been demonstrated to be sufficient to differentiate between it and other similar spindle cell or malignant fibrous lesions [3]. We suggest that if the lesion is small and amenable to excisional biopsy, then this is the treatment of choice. If the lesion is large or deep seated, then an incisional biopsy with a large and deep sample should be undertaken. Definitive treatment is wide local excision and reconstruction of the involved structures as needed.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Conflict of interest

There are no disclosures or conflict of interest to disclose.

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